Surgical Management of a Coronary-Bronchial Artery Fistula Combined with Myocardial Ischemia Revealed by $^{13}$N-Ammonia Positron Emission Tomography

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A 71-year-old male with known bronchiectasis and atrial fibrillation was admitted to Seoul St. Mary's Hospital with recurrent transient ischemic attack. Radiofrequency ablation was performed to resolve the patient's atrial fibrillation, but failed. However, a fistula between the left circumflex artery and the bilateral bronchial arteries was found on computed tomography. Fistula ligation and a left-side maze operation were planned due to his recurrent symptom of dizziness, and these procedures were successfully performed. After the operation, the fistula was completely divided and no recurrence of atrial fibrillation took place. A coronary-bronchial artery fistula is a rare anomaly, and can be safely treated by surgical repair.

Key words: 1. Coronary artery disease 2. Fistula 3. Bronchial arteries

Case report

A 71-year-old male with known atrial fibrillation, diagnosed 2 years previously, visited Seoul St. Mary's Hospital with symptoms of dysarthria and motor weakness. The patient had a history of lobectomy for treating uncontrolled bronchiectasis in his 30s. The symptoms were eliminated soon after hospitalization without any intervention, and the patient was discharged with the diagnosis of a transient ischemic attack. However, the symptom of dysarthria took place again. Based on the possibility of thromboembolism of the cerebral arteries, which can be caused by atrial fibrillation, radiofrequency catheter ablation was performed to treat his atrial fibrillation, but unfortunately his cardiac rhythm did not convert to sinus rhythm. However, in a multidetector computed tomography (MDCT) scan that was taken to verify whether coronary artery disease was present before the ablation procedure, an abnormal communication between the left circumflex artery (LCX) and the bronchial artery was discovered incidentally (Fig. 1A). The coronary arteries themselves were revealed to be intact. With the suspicion of a relationship between those symptoms and the coronary-bronchial artery fistula, which could induce myocardial ischemia, a gated $^{13}$N-ammonia positron emission tomography (PET)–computed tomography scan with an adenosine stress test was performed, and showed global left ventricular ischemia in the resting state.
Cardiac contractility likewise did not sufficiently increase after adenosine administration (Fig. 2A, B). The flow rates at rest were 0.78 mL/g/min for the left anterior descending artery (LAD), 0.78 mL/g/min for the LCX branch, and 0.66 mL/g/min for the right coronary artery (RCA). The flow rates in the adenosine stress were 1.06 mL/g/min for the LAD, 0.98 mL/g/min for the LCX, and 0.84 mL/g/min for the RCA. The coronary flow reserve was 1.37 mL/g/min, 1.24 mL/g/min, and 1.27 mL/g/min, respectively, and the average flow reserve was 1.31 mL/g/min, which indicated a generalized decrease of the coronary flow reserve, suggesting the possibility of diffuse microvascular disease (Fig. 2C). The flow reserve in the LCX territory was lower than that of the other areas, but the decrease in flow was not espe-
specially significant. Transthoracic echocardiography revealed only biatrial enlargement without a wall motion abnormality, and no coronary artery disease was found on a coronary angiogram (CAG). A large fistula from the LCX to both bronchial arteries was present on the CAG (Fig. 1B, C), as previously shown on the MDCT scan.

In order to resolve the patient’s symptoms, closure of the coronary-bronchial artery fistula was scheduled. Since pharmacologic and endovascular management of the patient’s atrial fibrillation had failed before, both the surgical ligation of the fistula and a maze operation via median sternotomy were planned. After median sternotomy followed by pericardiotomy, a tortuous fistulous tract with a diameter of approximately 1.5 mm was found on the roof of the left atrium. After cardiopulmonary bypass with cardioplegia, dissection and clipping of the fistulous tract were performed. A left-side maze operation was also conducted with a cryocatheter, and internal obliteration of the left atrial appendage was performed with a 4-0 prolene suture. Electrocardiography after surgery showed a regular sinus rhythm without recurrence of atrial fibrillation, and a follow-up CAG revealed the successful occlusion of the coronary-bronchial artery fistula (Fig. 3). The patient was discharged with an uneventful postoperative recovery, and no symptoms occurred within the 10-month follow-up period.

Discussion

Coronary-bronchial artery fistula, an abnormal communication between the coronary arteries and the unilateral or bilateral bronchial arteries, is a rare anomaly that is present in only 0.5% of patients who undergo coronary angiography [1]. According to a review article from the Netherlands, only 31 such fistulas were reported in the period from 2008 to 2013 [2]. Due to the advanced radiologic techniques that are currently used, such as MDCT, diagnosing coronary-bronchial artery fistula has become less complicated [3,4]. However, the etiology of coronary-bronchial artery fistula is uncertain. Said et al. [2] suggested the possibility that they involve the reopening of preexisting, nonfunctional congenital communications between the bronchial arteries and the coronary arteries. They proposed that 2 factors regulate the reopening and growth of arterial communications: disequilibrium of the pressure gradient between the 2 arteries and obstruction of the coronary arteries. Additionally, several case series have implied the possibility of a relationship between coronary-bronchial artery fistula and known bronchiectasis [2,5]. Similar to the patients who have previously been described, our patient also suffered from bronchiectasis. The progression of bronchiectasis leads to hypertrophy of the bronchial arteries, and this change in the bronchial arteries may influence the presence of communication between the coronary arteries and the bronchial arteries. Further studies are needed to understand the etiology of coronary-bronchial artery fistula and its relationship with bronchiectasis.

The clinical presentation of patients with coronary-bronchial artery fistula depends on the degree of the left-to-right shunt and the concomitant disease process in the patients. Said et al. [2] reported that chest pain was the most frequent symptom (63%) in their review, and Lee et al. [4] suggested an association between the coronary steal phenomenon and chest pain in coronary-bronchial artery fistula patients. Hemoptysis (26%) and dyspnea (19%) are also frequent, and otherwise asymptomatic disease occurred in only 5 of 27 subjects (19%) [2]. The patient in our report complained of recurrent dizziness and syncope, but not of chest pain or hemoptysis. However, we cannot infer that the patient’s symptoms
were due to the fistula, because they could also have arisen from underlying atrial fibrillation.

Although coronary angiography has emerged as the preferred diagnostic modality for coronary-bronchial artery fistula, the invasiveness of this procedure is a major obstacle. Noninvasive contrast-enhanced MDCT is as useful as CAG for diagnosing coronary-bronchial artery fistula and identifying the course of a fistulous tract [4,6]. According to the reviews by Lee et al. [4] and Said et al. [2], coronary-bronchial artery fistula originated in the circumflex artery in 75% (6 of 8) and 61% (19 of 27) of cases, respectively. Transthoracic or transesophageal echocardiography is also helpful in detecting coexisting cardiac anomalies and assessing the cardiac function of the patient. PET using 13N-ammonia, which was performed in this case, is also useful for the assessment of cardiac function. Quantification of absolute coronary flow and measurement of the coronary flow reserve by 13N-ammonia PET have the advantage of identifying the diseased vessel, and these techniques can also be valuable in assessing coronary-bronchial artery fistula [7].

Since the appropriate treatment modality for coronary-bronchial artery fistula has not been established, the fistula should be treated if a patient shows symptoms, in order to prevent lethal complications such as myocardial infarction, infective endocarditis, and aneurysmal rupture. Percutaneous transcatheter embolization may be the treatment of choice in most patients without concomitant cardiac disease, whereas surgical ligation is also effective in selected patients [2]. Patients with coronary-bronchial artery fistula and concomitant cardiac disease need more consideration for selecting the best treatment modality. In our case, the patient experienced atrial fibrillation as well as coronary-bronchial artery fistula. To manage both of these diseases at once, surgical ablation and fistula revision under median sternotomy followed by cardiopulmonary bypass was performed, and the coronary-bronchial artery fistula was successfully repaired.

**Conflict of interest**

No potential conflict of interest relevant to this article was reported.

**References**